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## **Adaptive seating and adaptive riding in children with cerebral palsy**

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Chapter 6

## General discussion



The main aim of this thesis was to increase knowledge about two types of postural control intervention in children with CP. The first intervention focusses on the optimal postural support provided by adaptive seating. The second intervention addresses and explores the training of postural control by an adaptive horseback riding program (TDAR).

Our studies, as discussed in this thesis, were undertaken to assess the effects of intervention across all the domains of the ICF-CY framework (WHO, 2007). The discussion is presented in three parts:

**Part I** addresses the effect of seating inclination and foot-support in ambulatory children with CP functioning at GMFCS level I-III during sitting while reaching; outcome was only assessed in the domain of body structure and function;

**Part II** discusses the effects of adaptive seating systems (AdSS) in children with severe CP, functioning at GMFCS level IV or V in all the domains of the ICF-CY;

**Part III** addresses adaptive horseback riding (TDAR); outcome was addressed in the domains of body structure and function and activities.

First, I elaborate on the three themes mentioned above, next I address methodological issues, while finally I explore the general clinical implications, offer recommendations for further research, and end with concluding remarks.

## Part I

**Chapters 2 and 3** evaluated the effect of adaptive seating interventions (15° forward (FW) tilting of the seat-surface and foot-support) on a set of outcome measures, including postural control and reaching quality during sitting while reaching of ambulatory children with spastic CP (US-CP and BS-CP, GMFCS levels I-III). A previous study had addressed the effect of the 15° forward tilting of the seat surface in the absence of foot-support.<sup>1</sup> It had reported that children with US-CP benefitted from forward tilting, but children with BS-CP did not. To our knowledge, no studies have focused specifically on foot-support as a possible factor affecting postural muscular adjustments during forward reaching tasks while sitting. We carried out a cross-sectional study using a repeated-measures design and addressed the following questions: does FW-tilting or horizontal seating, with or without foot-support affect the kinematics of head stability and reaching, and EMG-parameters of postural control during reaching? In particular, we were interested in the effect of foot-support in both tilting conditions. In **Chapter 2**, we had hypothesized that the provision of foot-support may result in a better head stability, i.e. a smaller angular sway of the head<sup>2</sup> and a better reaching quality, e.g. larger transport MUs. In **Chapter 3**, the effect of the seating interventions on postural muscle activity was studied at both levels of postural control. We had hypothesized that the potential effect of additional foot-support was best expressed in the second level control, i.e., in the child's ability to modulate EMG-amplitudes at baseline (the tonic background activity of postural muscles) and during reaching. Especially, we evaluated whether

the seating condition affected the correlations between the capacity to modulate EMG-amplitudes and the kinematics of reaching and head stability. During all data analyses, attention was paid to the putative effects of type of CP (US-CP or BS-CP) and severity of CP expressed by the child's GMFCS level.

The data revealed that seating condition did not affect the kinematic parameters of head stability during reaching. Yet, our findings confirmed the differential effects of seating condition on the kinematic reaching quality described previously by Hadders-Algra et al. (2007).<sup>1</sup> They reported that children with US-CP benefitted from 15° FW-tilting, while in children with BS-CP the FW-tilting worsened the quality of reaching. The present study showed that these effects were independent of foot-support. The better reaching quality was reflected by a reduction in the number of MUs and an increase of the transport MU. These findings suggest that children with US-CP have a larger access to feedforward (pre-programming) movement control in the 15° FW-tilting condition, whereas children with BS-CP can integrate more feedforward control in the horizontal seating condition. The FW-tilted condition was especially difficult for the children with BS-CP when foot-support was added; our data indicated that the addition of foot-support was associated with an increased duration and pathway of the reaching movements. This suggests that these children needed most feedback corrections in this situation compared to the other sitting conditions.<sup>1,3,4</sup> This also suggests that the children needed least feedback corrections in their optimal seating condition.<sup>3,4</sup> Shifting the focus from less feedback control to more feedforward control has the advantage that the child with CP does not need to spend much attention and cerebral effort on the task at hand (here: reaching while sitting), but can devote more attention and cerebral effort to the environmental context, including its cognitive<sup>5,6</sup> and language and interactive aspects.<sup>5</sup>

The provision of foot-support – our specific interest – was in all children with spastic CP associated with increased reaching velocity, increased baseline EMG-amplitudes and decreased phasic EMG-amplitudes of the trunk extensors, irrespective of seating inclination. The improved baseline EMG-amplitudes were – in turn – associated with better kinematics of reaching, i.e., increases in the transport MU – as mentioned before, one of the main parameters reflecting feedforward control of reaching. These findings suggest that foot-support may promote postural fine-tuning at the second level of postural control. This corresponds to the suggestions provided by Myhr et al. (1995)<sup>7</sup> and Hadders-Algra et al. (2007).<sup>1</sup> Their studies did, however, not provide evidence for the effect of foot-support.

The effect of the seating intervention did not depend on severity of CP (GMFCS levels I to III). This non-effect of GMFCS scaling was consistent with another study on postural adjustments in sitting in children with GMFCS level I-III.<sup>8</sup> However, this should be interpreted with caution, as the severity subgroups were small in our study. Importantly, the proportion GMFCS-level I school-age participants in our study was relatively high (12/19), which may have obscured a potential severity effect. Nevertheless, the relatively high proportion corresponds to the prevalence of GMFCS-level I reported in the literature.<sup>9,10</sup>

Overall, the data from **Chapter 2 and 3** suggest that optimal seating for children with US-CP consists of FW-tilting with foot-support, that for children with BS-CP of a horizontal seat surface with foot-support. The better reaches in the foot-support condition is, in part, enhanced

by an improvement of the children's capacity to modulate trunk extensor activity. It should be realized that only children with mild to moderate forms of CP have been studied (GMFCS levels I to III) were included in the study. In these children, the effect of the seating intervention did not depend on the severity of CP.

## Part II

AdSS intervention is part of postural management programs for children with severe CP, functioning at the GMFCS levels IV or V.<sup>11</sup> Its aim is to encourage children to engage in meaningful participation in daily activities. In the ICF-CY model, AdSS intervention is viewed as the immediate environment<sup>9</sup> of the child and may act to facilitate change in the child's functioning across all levels of the ICF-CY model.<sup>12</sup> Theoretically, AdSS that include hip- or pelvis-stabilizing devices and trunk support devices are regarded as essential in the achievement of a 'functional sitting posture'.<sup>13–15</sup> Therefore, in **Chapter 4** we addressed the following questions: what are the effects of specific components of AdSS, i.e., hip and pelvis stabilizing devices and trunk support devices on the child's function, specified for the three domains of the ICF-CY: Impairment, Activity and Participation? In addition, we assessed whether the functional effect of AdSSs depends on the severity of CP (GMFCS level IV or V) or the type of CP?

The analyses revealed that only nine studies had sufficient methodological quality to be included in the final analyses.<sup>16–24</sup> Nevertheless, it should be realized that the nine studies only had level IV evidence and were of moderate methodological quality. The outcomes reported in the nine best studies were divided into three categories: postural function, upper extremity function, and additional outcomes by means of activity and participation. However, most of the studies ( $n = 7$ )<sup>16–18,20–23</sup> addressed outcomes only in terms of postural function. Five of them did not find a significant improvement of the AdSS on postural function. Two of them did.<sup>20,21</sup> These studies may provide us with cautious suggestions about what might be effective AdSS to improve the child's postural function. In terms of 'Body Functions and Structures', the effect of trunk- and hip-support devices may improve postural alignment and the postural control outcomes.<sup>20,21</sup> The devices for which a beneficial effect was shown were a) the CAP-II seating system that improved symmetrical trunk posture of children with scoliosis<sup>20</sup>, and b) the X-PANDA seating system in combination with a dynamic backrest that improved postural stability and upper extremity function in children with dyskinetic CP.<sup>21</sup> The latter group of children often suffers from extensor thrusts (involuntary high-intensity muscle contractions) that induces a non-functional extended posture.<sup>21</sup> The postural control studies indicated that in children functioning at GMFCS level IV-V the basic level of control is present,—be it sometimes with impairments—but that they have serious impairments to adapt their posture to the context.<sup>14,25</sup> Therefore, from a theoretical point of view, it is conceivable that in these children provision of postural support devices, that support the child at the hip, pelvis and trunk, reduces the number of degrees of freedom which have to be controlled. This will reduce the demand on postural control and its associated upper extremity task.<sup>14</sup> This was also shown by the study of Cimolin et al.<sup>21</sup> which was the only one out of

four studies that addressed outcomes on upper extremity function. Cimolin et al.<sup>21</sup> indicated that a dynamic back rest is associated with a reduction of extensor thrust activity, i.e., better head and trunk stability, which in turn decreased involuntary arm movements.<sup>1</sup>

The studies included in the review mostly evaluated AdSS devices that included support-components at the level of the foot, pelvis, and trunk. The effect of support at the level of the neck was infrequently studied. This is at variance with clinical practice in which children with severe forms of CP virtually always receive devices supporting neck and head. This means that the effect of neck and head support needs further evaluation. Another concern is that most studies evaluated short-lasting effects in an experimental set-up. It has been suggested that short-lasting effects on the child's postural impairments may differ from the long-lasting effects on the child's performance in daily life.<sup>15,26,27</sup> However, this hypothesis has never been tested. One aspect of the long-lasting use over the day of a specific AdSS is the issue of seating comfort, especially in children with CP who have severe communicative and cognitive impairments. For example, children functioning at GMFCS levels IV or V, with scoliosis, might need a postural support device counteracting the asymmetry of the scoliosis during seating, but this specific AdSS<sup>20</sup> may apply uneven pressure distribution on the skin and otherwise may also cause stress and pain due to the forces induced by the AdSS. Unfortunately, none of the studies in **Chapter 4** addressed this outcome. Nevertheless, in none of the studies adverse side effects of the AdSS were reported. Yet, caution is warranted. This means that future studies should take this into account. They could, for instance, prospectively use the CP-Child questionnaire (CPCHILD)<sup>28</sup> to holistically evaluate comfort or pain level, achievement of functional activities and participation of children in different settings, e.g. home, school, or community during AdSS-use.

The studies addressing outcomes in terms of 'Activity' dealt with prehension, self-care and play. The specific-purpose AdSS, i.e. the Flip2Sit activity seat and the Aquanaut toilet system, were associated with improvements of these daily life activities.<sup>19, 24</sup> The parents reported that these AdSS also improved the children's participation at home, at school and in the community.<sup>19,24</sup> These findings are encouraging. The findings above illustrate that intervention using AdSS may be viewed as influencing the 'immediate environment' of a child and that their application may act as a facilitator for change in function in all domains of the ICF-CY.<sup>9,12</sup> Therefore, I suggest that future research applies a battery of tools to assess the effect of AdSS on daily life activities, including the assessment of family wishes and burden.<sup>12,26,29</sup>

Previously it has been suggested that the putative effect of AdSS depends on the severity of CP.<sup>2,14</sup> However, we were unable to identify an effect of the child's CP type and severity level (GMFCS level IV vs. V) on the outcomes achieved by AdSS intervention due to lacking details in the nine studies. Finally, eight out of the nine studies dealt with school-age children, with only five<sup>18,19,22-24</sup> of the studies addressing the effect of AdSS in some children younger than six years of age. The latter meant that we could not evaluate whether the effect in the younger children differed from that in the school-age children. It is, however, conceivable that the effect of AdSS at

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1 Note that an error occurred in Chapter 4; we erroneously reported there that the AdSS in the Cimolin et al. study was associated with an adverse effect of arm movements. However, the effect was favorable.

an early age is greater than that at later ages<sup>11,30,31</sup>, as the nervous system has greater plasticity at the early age (Hadders-Algra, 2014).<sup>32</sup> Thus, I recommend that future studies in particular address the effect of AdSS in young children with severe CP.

## Part III

In **Chapter 5**, we indicated that it is technically and physically feasible to perform our TDAR-intervention study in a relatively short period of time, as the intervention only lasted 6 weeks. All children participating were enthusiastic and fully engaged in the TDAR program. The study indicated that all children with spastic CP functioning at GMFCS level III were able to participate both in the TDAR-intervention and in its evaluation.

The study's secondary aim was to evaluate the effect of TDAR-intervention. The TDAR consisted of six weeks applied twice per week for one hour. We hypothesized that TDAR-intervention would be associated with a significant improvement of gross motor function, especially for the dimensions D (standing) and E (walking, running, jumping) of the GMFM-88. The results suggested indeed that the TDAR-intervention was associated with an improvement in GMFM dimensions D and E that exceeded the minimally clinical important differences (MCIDs). The latter suggests that the changes were clinically meaningful.<sup>33</sup>

Another potential effect was expressed in terms of reduced stereotypical myographically recorded postural adjustments during sitting while reaching. We also found that at the end of the intervention, fewer children were accompanied by a side walker during TDAR which may suggest that their riding skills improved. This suggestion was supported by the videos of the TDAR intervention that indicated that the children could better cope with postural challenging exercises. Our study suggests that prolonged practice of postural challenging tasks may improve children's motor function and postural control. We suggest that the following TDAR ingredients contributed to its favorable effect. Firstly, the hands-off approach forced the children to self-practice and finding their optimal strategies by trial and error. Secondly, children were exposed to different experiences, as each child experienced six different horses, each with different characteristics. Thirdly, the integrated program of varied postural challenge exercises was an essential component of TDAR.

Nevertheless, our TDAR program did not result in significant changes in the secondary outcome measures at other levels of the ICF-CY. This is similar to the findings in the larger study of Davis et al. (2009).<sup>34</sup> Yet, we feel that it is too early to conclude that the TDAR program had no effect on the children's Activity and Participation levels, as our study lacked the power to demonstrate such an effect. Of course, the significant improvement in GMFM-88 scores in a small study group needs to be interpreted with caution. Importantly, the child's age is one potential and important factor that could affect the changes in the GMFM performance. In our study the two youngest children, aged 6 and 7 years, showed the largest changes both during the two-baseline testing (T0-T1) and during the intervention (T1-T2). The increase in age and its associated developmental changes may have acted as a confounder. The confounding effect may be two-fold. Firstly, children with CP gradually grow into their deficit, implying that function gets worse over



time. Secondly, the GMFM assessment may have a ceiling effect in older children; thus, it may not fully reveal all aspects of the developmental changes occurring in a child. These problems may be avoided by using the Goal Attainment Scaling that addresses the individual child's performance.

Finally, an important factor that may have explained the effect of our TDAR intervention may have been its intensity. It is getting increasingly clear that the intensity of therapy is a significant factor determining the outcomes of physical therapy in children with CP.<sup>35</sup> This may also be true for horseback riding interventions. The literature suggests that the studies that employed an intensive exercise program in horseback riding therapy or hippotherapy reported a positive effect on the Impairment and Activity domains of the ICF-CY.<sup>36–40</sup> In contrast, the studies that applied a less intensive therapy were associated with no changes in gross motor performance.<sup>41,42</sup>

In the following paragraphs I address methodological considerations and the limitations of the studies included in this thesis. I end with concluding remarks and perspectives for future research.

## **Methodological considerations**

### **Part I: seating inclination and foot-support**

The strength of the studies in Part I is, firstly, the standardized measurements in children with CP (who were randomly assessed in four seating conditions), i.e. the kinematic assessment of head sway and reaching, and the standardized measurement and analyses of the EMG-recordings of postural adjustments. A second strength is that the kinematic and EMG data were analysed by assessors who were masked with respect to seating conditions, therewith reducing the risk of bias. A third strength is group composition: the two subgroups studied (BS-CP and US-CP) were similar in their clinical characteristics. A fourth strength is the application of mixed-effect model analyses, allowing for the correction of the confounding effects of age and body proportions and taking into account the existing correlations in the data.

The studies also had some limitations. As mentioned above, our findings cannot be generalized to all children with CP, as we only studied ambulatory children with spastic CP and functioning at GMFCS levels I to III, within a specific age range (6–12 years), without significant co-morbidity. Second, the small sizes of the subgroups are additional limitations, implying that the analyses on the effect of severity of CP should be interpreted with caution. Especially the proportion GMFCS-level I participants was relatively high (12/19), which may have obscured a potential severity effect. Finally, we studied the effect of seating only once in a laboratory setting, which precludes direct generalization to a variety of activities in everyday environments.

### **PART II: Effect of AdSS across all the domains of the ICF-CY**

It is generally recognized that research in this area is very difficult.<sup>14,29</sup> This is reflected by the low grades of evidence of the nine methodologically best studies included in the review. This low grade of evidence, the moderate and varied quality of the methodological approach, and little

well-supported information are a serious limitation of our **Chapter 4**. However, we systematically reviewed the material available by adapting the standard systematic review protocol by using a triple methodological quality assessment<sup>43–45</sup>, in order to assess methodological quality and risk of bias. The detailed methodological analysis thus was regarded as the strength of the systematic review. The second strength of the review is its systematic and structured organization. We organized outcomes systematically, i.e., according to functional outcomes (postural function, upper extremity function and additional outcomes) with special attention to three domains of the ICF-CY (WHO, 2007)<sup>12</sup> as a research framework. Before the year 2001, i.e. before the implementation of the ICF-CY<sup>12</sup>, research on AdSSs focused on the Impairment level (outcome measures in the BS/F domains). Only after the introduction of the ICF-CY attention in AdSS-research gradually shifted to the Activity level of the ICF-CY. However, studies addressing the Participation level are still rare. The net result for the review was that we had little information on the effect of AdSSs on Activity and Participation. Nevertheless, I think that the classification of the study outcomes in terms of ICF levels (BS/F, A, and P) is very useful in the communication between multidisciplinary health professionals. The information in **Chapter 4** can highlight essentialities: which AdSS interventions have the highest potential to maximize functioning, especially taking the Activity and Participation levels into account.

I like to stress again that the most serious limitation of the review is that it is based on studies with a low level of evidence. Another limitation is related to the previous limitation. We slightly adapted the criteria of the AACPD<sup>44</sup> score for moderate methodological quality in order to preserve a sufficient number of studies in the review. This is a choice that may be debated. Finally, the heterogeneity of the studies precluded the performance of a meta-analysis to balance overall outcomes across studies.

## PART III: TDAR study

The strength of the TDAR study is that the feasibility of a complex study protocol was established. Another strength is the use of the MCID outcome tool of the GMFM-88 score for ambulatory children with CP. The MCID is the magnitude of change required for determining when a meaningful change occurs. If a change score exceeds MCID, it is likely that change is of clinical importance.<sup>33</sup> Thus, the MCID results of the TDAR study suggested that the statistically significant change in GMFM-88 scores was clinically relevant. Application of the MCID also assists in power calculations and in tool selection for future studies.<sup>33,46</sup> Other strengths are our well specified TDAR training protocol including novel and promising strategies applied during riding activities. A final strength is the application of the standardized video analysis specifically designed for behavioral observation, in the TDAR study adapted for the analysis of the events during TDAR.

The limitations of our TDAR study are related to its design as feasibility study with a small sample of children who had CP and functioned at GMFCS level III. This means that the design does not allow for generalization to all children with CP. In addition, the use of only one post-intervention assessment precluded conclusions about long-term effects. Another limitation was that the assessors were not masked with regard to the intervention; this limitation posed the risk

of detection bias.<sup>47</sup> The exception to this rule was that the assessor of the EMG-data was masked. Finally, it may be regarded as a limitation that we performed our TDAR-video analysis only twice, i.e., in a minority of TDAR sessions.

## Clinical implications

### Seating inclination and foot-support for ambulatory children with CP

An important finding of our studies was that the optimal seating conditions for children with US-CP differed from those for children with BS-CP. Our findings suggested that in ambulatory children with US-CP FW-tilted seating with foot-support offers the best situation to achieve optimal reaching movements. In children with BS-CP, the horizontal seating with foot-support apparently is best. Our results on foot-support are in line with clinical suggestions about the effect of foot-support. The clinical idea is that in children with CP, appropriate foot-support during sitting reduces the degrees of freedom of the lower extremities, thereby enhancing stability during sitting.<sup>7,14,18</sup>

Our studies indicated that the optimal seating condition was associated with more feed-forward control of the reaching arm. The latter suggests that the optimal seating condition may help the children with CP to spend not too much concentration and cerebral effort on the programming of the arm movements. Instead, more attention and cerebral effort may be devoted to the environmental context, including its cognitive<sup>5,6</sup>, language and interactive aspects.<sup>5</sup> Whatever limitations may be perceived in our study, I am confident that our results provide physiological guidance for, and may well inform clinical decision making by clinicians and therapists who are involved in adaptive seating interventions in children with CP.

Nevertheless, we also found that children's head sway was not affected by seating conditions. Thus, in children with GMFCS level I-III, in whom head stability is rather well controlled, the effect of optimal seating position is often expressed in better upper-arm function.<sup>1,48,49</sup> In children with severe CP the situation may be different. For instance, Hadders-Algra et al. (2007)<sup>1</sup> found that the horizontal seating condition was optimal for children with BS-CP, including children functioning at GMFCS level IV. This may suggest that the horizontal seating condition serves better head control in children with more severe forms of BS-CP.

### AdSS for children with severely CP: in the context of ICF-CY perspectives

The systematic review suggested that trunk and hip support devices possibly may improve postural alignment and postural control of children with severe CP functioning at GMFCS level IV to V.<sup>20,21</sup> However, we increasingly realize that – in line with the ICF-CY – the effect of AdSSs on 'Activity and Participation' is more relevant for child and family. Currently our knowledge on such effects of AdSS is very limited. Nevertheless, the review suggested that AdSS in the form of 'Flip2sit' activity seat and 'Aquanaat' home-toilet seat may be able to improve activities in daily life, including self-care and play.<sup>19,24</sup> Conceivably, the potentially beneficial effects of AdSSs that provide hip-,

pelvis-, and trunk-support in children with severe CP, is mediated by a reduction in the number of degrees of freedom which have to be controlled, thereby reducing the demands on the postural control task.<sup>14</sup>

I also would like to stress that the international guideline on 24-hour postural management in children with severe neuro-disability recommends that special seating should be introduced at six months of age.<sup>11</sup> Thus, I suggest that AdSS intervention in children with severe forms of CP – who may be diagnosed before the age of 6 months<sup>50</sup> – starts at a very early age.

## TDAR intervention in children with CP, GMFCS level III

Nowadays, a popular form of postural training in children with CP is therapy that uses horseback riding. Our findings (**Chapter 5**) suggested that the TDAR-intervention of six weeks, with an intensity of one hour, twice per week, including postural challenge exercises on the horse, may enhance gross motor function and postural adjustments in children with CP with ambulatory difficulties. For our TDAR, we opted for a group approach as it promotes active participation<sup>51–53</sup>, i.e., self-practice, minimization of therapist's hands-on guidance, and socialization within the group exercise. The latter is, in general, associated with an increase in the child's pleasure.<sup>12,54</sup> Clinically, the strategies used in TDAR postural training can be easily implemented. TDAR used the coaching of the riding instructor and therapist as a means of guidance of a child to perform the designed activities. Coaching is one of the novel and promising strategies used in pediatric physical therapy and rehabilitation.<sup>55</sup> Our TDAR coaching strategy emphasized the hands-off technique during riding sessions so that the child is given opportunities to explore his/her postural capacities.<sup>55</sup> However, I would like to emphasize that the feasibility study generated suggestions, not evidence.

## Future perspectives

In the field of seating technology, the nature of the best seating condition in children with CP is an ongoing debated issue. The debate is largely due to conflicting study results. The latter is presumably caused by the mixed composition of the study groups, the heterogeneity in seating conditions and measurement methods.

I suggest that future research on the effect of forward-tilt seating and foot-support should aim for replication in larger groups with equally sized subgroups per GMFCS level and including the measurements of performance outcomes across all domains of the ICF-CY.<sup>12,15</sup> In addition, future study may include more challenging reaching tasks, such as reaching for a target beyond arm length distance or in a variety of reaching directions in the context of everyday environments, such as during home and school tasks.

Future studies on the effect of AdSS in children with severe CP need to have a high methodological quality and preferably should address the effect on activity and participation. The latter two domains are – as the ICF-CY framework highlights – strongly dependent on the child's context, such as the family and school.<sup>12,26,29</sup> I suggest that future studies use a battery of tools to assess the effect of AdSS on daily life activities, including the assessment of family wishes and burden.<sup>12,26,29</sup>

Importantly, the evaluation of participation in children with severe CP is not possible in a laboratory setting.<sup>26,30</sup> The best options available are questionnaires and interviews. In young children and in children with limited communicative ability, a proxy respondent, e.g. the primary caregiver, is an acceptable alternative to the child.<sup>12,56,57</sup> The tools available are the COPM<sup>58,59</sup>, the GAS<sup>60,61</sup>, the Caregiver Priorities & Child Health Index of life with Disabilities (CPCHILD) questionnaire<sup>28</sup>, and the Family Impact of Assistive Technology Scale for Adaptive Seating (FIAT-AS).<sup>57</sup>

I also suggest that the studies on AdSS evaluate the comfort perceived in a specific AdSS. Various research strategies may be applied here, including the use of long-lasting (e.g. an entire day) pressure-distribution measurements, or the use of the CPOCHILD questionnaire to evaluate the child's comfort, the ease of care, health and quality of life.<sup>28</sup> Next, the qualitative video analysis<sup>53</sup> with masked assessors for type of intervention may be used to evaluate the child's non-verbal behavior, using scoring systems similar to the ones used in the evaluation of pain in newborn infants.<sup>27</sup>

Suggestions for future study groups may include but may not be limited to multicentre trials to increase group size, thereby allowing for adjustment for confounding by co-morbidities. Secondly, in this difficult area of research, prospective cohort design, mixed-methods design combining quantitative and qualitative approaches and high-quality single-subject research methodology presumably are the most rigorous designs. Detailed methods should include severity and document type of CP with standardized instruments, such as GMFCS and the guidelines of the Surveillance of Cerebral Palsy in Europe for CP classification (SCPE, 2000).<sup>62</sup> Importantly, because AdSSs for the most severely affected children is needed at an early age<sup>11</sup>, the effect of AdSS intervention in infancy and preschool age could be a research theme for future study.

Lastly, I suggest that future studies on "TDAR intervention" should use a randomized controlled trial design to compare outcome of intervention with that of non-riding controls, which utilizes a similar set of assessment tools in all participants, i.e., a battery evaluating outcome across the levels of the ICF-CY<sup>12,51</sup> – and if possible – assessment of working mechanisms. Ideally, postural control should also be assessed during the horseback riding intervention, future studies may embark on this endeavor. Next, relatively large numbers are needed as CP is characterized by heterogeneity (SCPE, 2000).<sup>62</sup> I recommend block-randomization for severity of CP (GMFCS I-III versus GMFCS IV-V). Ideally, the study should be so large that it has a sufficient number of children in the following age groups: preschool-age, school-age, and adolescents. I suggest that TDAR lasts for 3 months (1 hour, 2x per week), and that evaluation (carried out by masked assessors) includes follow-up at least 3 months post-intervention.

## Conclusion

### **The results of this thesis suggest the following:**

1. Children with US-CP, GMFCS levels I-III, benefit in terms of reaching movements most from FW-tilted seating with foot-support; in children with BS-CP, GMFCS levels I-III, the horizontal seating presumably is best, with a potentially minor positive effect of foot-support.
2. Adaptive seating systems constitute important assistive devices in the daily life of children with severe CP, GMFCS IV-V. Our systematic review concluded that the low level of evidence of available studies precluded pertinent conclusions on effectiveness of AdSS in children with severe CP. The review suggested that in particular special-purpose AdSS may have the potential to improve activities and participation.
3. The feasibility study on six weeks of TDAR intervention, provided as a twice weekly one-hour group session, including postural challenge exercises on the horse, showed that it is well feasible to perform a RCT using this complex protocol. In addition, the study suggested that this TDAR intervention may enhance gross motor function and postural adjustments in children with CP with ambulatory difficulties (GMFCS level III).

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